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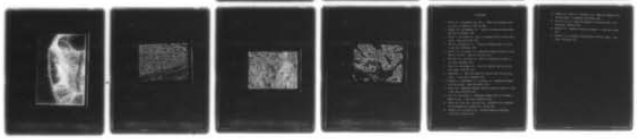

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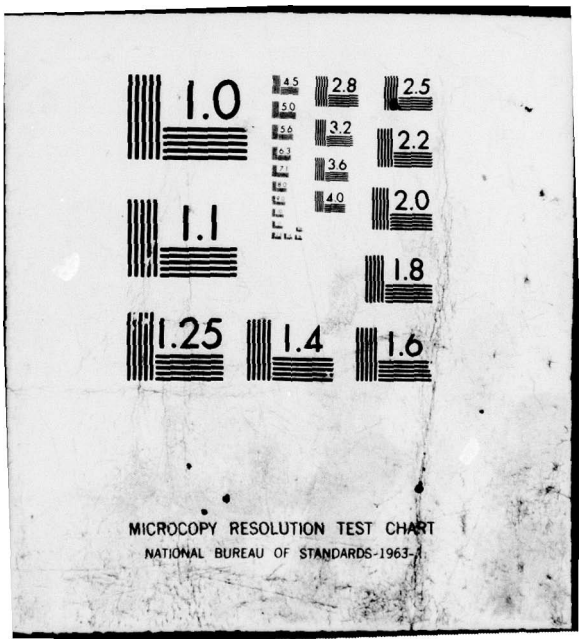
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A case report of an intraoral monomorphic adenoma is presented and the literature reviewed. The clinical manifestations, histology and treatment of this rare salivary gland tumor are discussed. Important clinical diagnostic points are stressed and the authors suggest this tumor be referred to as basaloid monomorphic adenoma.

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Abstract

A case report of an intraoral monomorphic adenoma is presented and the literature reviewed. The clinical manifestations, histology and treatment of this rare salivary gland tumor are discussed. Important clinical diagnostic points are stressed and the authors suggest this tumor be referred to as basaloid monomorphic adenoma.

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Intraoral Monomorphic Adenoma

by

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and

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Tumors of the salivary glands are uncommon despite the fact that many different types of tumors may affect these tissues. Salivary gland tumors constitute approximately 1-4 percent of all neoplasms of the head and neck.[1] One of the most uncommon of these tumors is the monomorphic adenoma which is composed of basaloid type cells that are arranged in various configurations including solid, cystic, tubular, trabecular and canalicular patterns. This tumor entity has been referred to by many names including: canalicular adenoma; monomorphic adenoma (canalicular type); monomorphic adenoma, canalicular variant; monomorphic adenoma, trabecular-tubular, canalicular and basaloid variants; monomorphic adenoma (basal cell adenoma); basal-cell adenoma; solid adenoma; trabecular adenoma and basophilic adenoma.[1-11]

This lesion has received attention in the recent literature.[3-11] The clinical findings in the reported cases have been remarkably consistent. The following clinical statistics are based upon review of 49 intraoral cases of this tumor (Table 1). The tumors were distributed as follows; 36 in the upper lip (frequently near the midline); 3 were "closely adjacent" to the upper lip; 2 in the lower lip; 1 in the lip (specific lip not mentioned); 5 on the palate (4 on the hard palate); and 2 on the buccal mucosa. Clinically, the tumor was characteristically described as a non-ulcerated, freely moveable mass which was seldom larger that 2.0 cm in its greatest dimension. Symptoms, when recorded, included: presence of a swelling, an increasing swelling, presence of a swelling that was "bothersome" or "uncomfortable" and swelling

which annoyed the patient. Pain, either spontaneous or with palpation, was not reported in any case. Nelson and Jacoway[5] stated that the lesion was painless in all of their 29 cases. The age of the patient was mentioned in 41 of the total 49 cases. Nelson and Jacoway[5] reported the median age of their 29 cases (which included one parotid lesion) to be 60.0 years. The average age of 12 other patients in which the age was recorded was 69.3 years. The sex of the patient was stated in 43 cases with 22 being male and 21 female. The race of the patient was recorded in 39 cases. Interestingly, 38 of the 39 cases occurred in Caucasians and the case report by Weiner[10] is the only case we are aware of in which this tumor occurred intraorally (minor salivary gland location) in a Black patient.

Treatment consisted of complete surgical excision in all cases. Recurrence was not observed in any case with follow-up periods which ranged from 2 months to 10 years and 8 months.[5,8]

Case Report

A 71-year-old Caucasian female was seen by the Oral Medicine Clinic of the Indiana University School of Dentistry for evaluation of a swelling under her upper lip which caused discomfort in the wearing of her complete maxillary denture. Clinical examination revealed a purplish mass located just to the left of the anterior maxillary frenum in the depth of the mucobuccal fold (Fig 1). The lesion was approximately 1.5 cm in diameter, covered with intact mucosa, freely movable, painless and not tender when palpated. The patient indicated that the enlargement had been present for "several months." She was referred to the Oral Surgery Department for excisional biopsy with the preoperative differential diagnosis of (1) salivary gland tumor (2) pyogenic granuloma (3) hemangioma.

The mass was excised and sent to the Oral Pathology Department for histopathologic analysis. The pathologist reported the lesion was composed of a well-defined mass of neoplastic epithelium dispersed in duct-like and cord-like structures which were compressed toward the periphery of the specimen (Fig 2). In the central portion of the mass these structures were widely separated and disrupted by large numbers of red blood cells located in vascular channels, organizing thrombi and mucoid material (Fig 3). Several areas toward the periphery were cylindromatous in configuration and exhibited multiple microcystic spaces. The neoplastic epithelial cells were cuboidal to columnar in shape and contained varying amounts of eosinophilic cytoplasm and prominent basophilic nuclei (Fig 4). Mitotic figures were not prominent. Abundant pale basophilic mucoid material was noted throughout the lesion separating cords of epithelium and filling the small cyst-like spaces. The mass was surrounded by a well-defined fibrous connective tissue capsule which was not violated by tumor cells. Diagnosis: Canalicular adenoma.

No recurrence of the tumor was detected at 3, 6 and 12 month follow-up examinations.

Discussion

After reviewing the literature concerning this tumor we feel that several important clinical characteristics should be emphasized as they will aid the clinician in the diagnosis of this uncommon lesion. First, it is interesting that several case reports, including the present one, state that the tumor was bluish or purplish in color.[6,7] It is also interesting to note that several other reports include "mucocele" as their provisional diagnosis or as part of their differential diagnosis.[3,11] While these reports do not specifically mention the color of the tumor, it would seem reasonable to

assume they may also have been bluish or purplish in color as this is one of the classical features of the mucocele of the lips.[12] We believe this color is commonly seen with the intraoral monomorphic adenoma and that the color of the tumor is a significant clinical finding which has not received adequate emphasis in the past. Several case reports, as well as the present one, displayed a prominent vascular component histologically. Christ and Crocker[3] emphasized the importance of the vascularity in making the histologic diagnosis of this tumor. They felt that the canalicular varient (apparently the most common variant) has a "pathognomonic vascular pattern in that small capillaries and venules predominate in the microcystic areas of the tumor." We suggest that this prominent vascular component accounts for the frequent clinical finding of a bluish or purplish color of the tumor especially when the tumor is located superfically. This color is an important point in arriving at the provisional clinical diagnosis as few other tumors present with this clinical appearance.

Another important clinical feature is the fact that the upper lip (especially the mid-portion) is by far the most common location for these intraoral monomorphic adenomas.[5] As indicated, review of the literature indicates that these tumors are frequently thought to represent mucoceles. However, the mucocele seldom occurs in the upper lip.[13] Standish and Shafer[13] reported no examples of mucous retention phenomenon (mucocele) of the upper lip in their series of 97 cases of this lesion.

The age of the patient is another significant factor to consider as this tumor almost always affects older people, usually between the ages of 60 and 80 years, as contrasted to the mucocele which usually occurs in the young.

A final point should be emphasized regarding the provisional diagnosis. Our literature review revealed only one case of this intraoral monomorphic

adenoma which occurred in a Black patient.[10] All of the other tumors occurred in Caucasians. The intraoral occurrence of this monomorphic adenoma in Blacks appears to be decidedly uncommon at the present time. Several examples of these tumors in the major salivary glands of Blacks have been reported, however.[14,15]

In summary, we feel that several clinical points should be emphasized as they are important in making the provisional diagnosis. These points are; the color of the tumor, frequently bluish or purplish; the tumor location, typically the middle third of the upper lip; the age of the patient, usually 60 to 80 years of age and the race of the patient, all reported intraoral cases except one occurred in Caucasian patients.

Two additional points, and really the most crucial ones, remain to be discussed. First, these benign tumors may closely resemble the malignant adenoid cystic carcinoma histologically. In fact, at least one of these benign tumors has been misdiagnosed in the past as adenoid cystic carcinoma and the patient needlessly subjected to radical surgical procedures.[16] Secondly, we believe the clinician should question a pathology report of adenoid cystic carcinoma for a lesion which fulfills the clinical characteristics of the intraoral monomorphic adenoma. This opinion is based upon the fact that while these two tumors may closely resemble one another histologically, the treatment and prognosis of the two lesions is vastly and justifiably different. The clinical characteristics become important in this circumstance because while both the monomorphic adenoma and adenoid cystic carcinoma typically affect older people, adenoid cystic carcinoma seldom occurs in the upper lip, and seldom, if ever, has been reported as being bluish in color or freely moveable.

The suspicions of the clinician, based upon the clinical characteristics of the lesion, may save a patient needless radical surgery if a monomorphic adenoma should be histologically misdiagnosed as an adenoid cystic carcinoma and treatment determined on the microscopic findings alone.

The more than passing resemblance of this monomorphic adenoma to the adenoid cystic carcinoma of salivary gland origin is worthy of further mention. It is not unreasonable to speculate that the monomorphic adenoma may be the benign counterpart to this malignant tumor.[17] The present case presents with aspects at the microscopic level which add credance to this possibility. The individual basaloid cell type as well as the cylindromatous configurations along with the suggestion of a basophilic mucoid material in microcystic areas number among these.

One is faced with a dilemma in finally settling upon a name for these tumors. Is it sufficient to simply differentiate the single cell type of salivary gland adenoma (monomorphic) from the multiple cell type of tumor (pleomorphic)? It is the opinion of the authors on the basis of tumors actually studied and the histological descriptions of other reported cases that the tumor cells manifest as basaloid type cells in every case. Some tumors are more solid and cellular while others have a tubular, canalicular and even plexiform configuration. Is it truly necessary to further distinguish them if there is unanimity of opinion as to the constancy of the cell type itself? All epithelial elements of this tumor are distinguishable morphologically as basically adenoid in nature although the glandular pattern may vary, primarily with the cellularity. Confusion could ensue if one were to split hairs and consider, for example, the oncocytoma as a monomorphic adenoma since it is an adenoma of salivary gland origin and is unicellular in type. While no diagnostic problem exists between the oncocytoma and the

monomorphic adenoma it is nevertheless a nosological challenge. It is always possible, of course, that further case reports will reveal new cases which don't conform to the factors mentioned above. Until such a time, however, perhaps it would be appropriate to refer to this single cell-type of salivary gland adenoma simply as the basaloid monomorphic adenoma. The term "basaloid" describes the cell type, "monomorphic" the lack of diversity and "adenoma" as the benign glandular nature of this tumor. This terminology is in direct and convenient contrast to the pleomorphic adenoma of the salivary glands and yet provides an essential descriptive element. If further descriptions are desirable one could append the term basaloid monomorphic adenoma with the mention of the variant. An example, as applicable in the present report, would be basaloid monomorphic adenoma, canalicular variant.

SUMMARY

Clinical manifestations, histology and treatment of an uncommon salivary gland tumor are discussed. The name basaloid monomorphic adenoma is suggested for this tumor.

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Disclaimer

The opinions and assertions contained herein are the private views of the authors and are not to be construed as official or as reflecting the the views of the Department of the Army or the Department of Defense.

Biographical Sketch

Dr. Mader is the Assistant Chief of the Oral Medicine Service of the Walter Reed Army Medical Center, Washington, D.C. Dr. Nelson is the Chief of the Division of Pathology, The U.S. Army Institute of Dental Research, Washington, D.C.

Captions

Figure 1. Preoperative photograph of 71 year old female patient. Mass is apparant in the left anterior maxillary frenum area. Note vascularity.

Figure 2. Histologic section of monomorphic adenoma composed of duct - like and cord - like neoplastic epithelial structures compressed toward the tumor capsule. (Hematoxylin and eosin stain. Magnification, X 120)

Figure 3. Histologic section of central portion of tumor mass. Note vascular channels, red blood cell concentrations and foci of mucoid material. (Hematoxylin and eosin stain. Magnification, X 40)

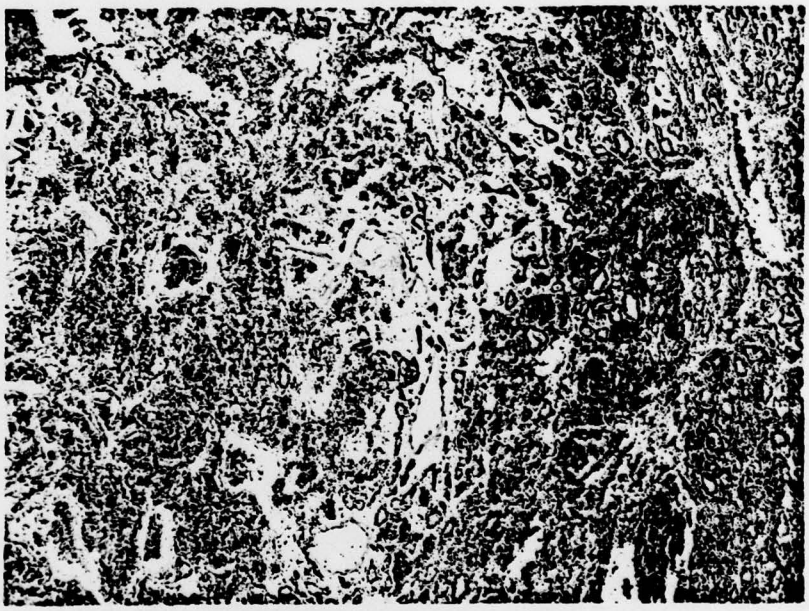
Figure 4. Histologic section showing cuboidal epithelial cells arranged in an adenomatous pattern located in a vascular connective tissue stroma. (Hematoxylin and eosin stain. Magnification X 300)

Table 1

Intraoral Basaloid Monomorphic Adenomas with Published Clinical Data

Author and Year	No. of Cases	Location	Age	Sex
Eggers-1928	1	palate	NA*	NA
Bhaskar & Weinmann 1955	4	upper lip lower lip hard palate cheek	NA NA NA NA	NA NA NA NA
Kleinsasser & Klein 1967	1	upper lip	65	M
Evans & Cruickshank 1970	2	lip upper lip	40 56	F F
Davis & Davis 1971	1	upper lip	87	M
Christ & Crocker 1972	3	upper lip upper lip upper lip	60 56 78	F M M
Nelson & Jacoway 1973	29**	23 upper lip 3 closely adjacent to upper lip 1 lower lip 1 hard palate 1 parotid area	Median age Was 60.0	16 M 13 F
Klein & Goldman 1973	2	upper lip buccal mucosa	59 74	M F
Bollinger & Hiatt 1973	1	upper lip	73	M
Strychalski 1974	1	upper lip	71	F
Crumpler, Scharfenberg & Reed 1976	2	hard palate hard palate	NA NA	NA NA
Wiener-1977	1	upper lip	67	F
Frim & Giunta 1979	1	upper lip	71	F
Mader & Nelson 1980	1	upper lip	71	F

* NA - information not available in report
** One tumor was located in the parotid area



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